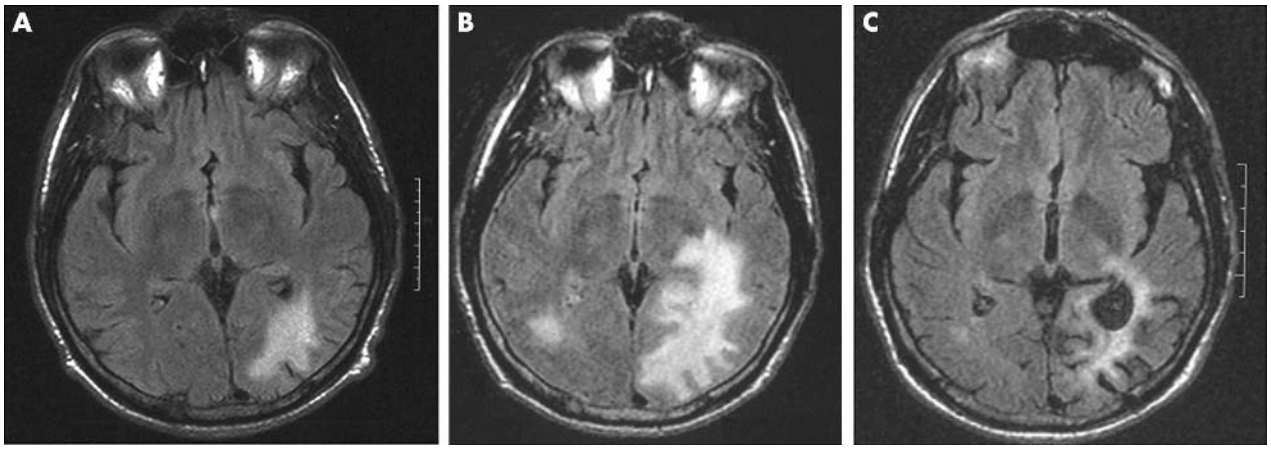


**Figure 1** evolution of cerebral magnetic resonance imaging (MRI) before and during treatment. (A) The first cerebral MRI, two months after the first neurological symptoms. Flair weighted axial images (TR 9000 ms; TE 114 ms) showing high signal intensity lesion in the left occipital white matter with respect to cortical grey matter. (B) Brain MRI one week after treatment onset. Flair weighted axial images (TR 10 002 ms; TE 160 ms) showing extension of the lesions. At that time, extension of the lesions was greater, with bilateralisation of the lesions in both occipital white matter and extension through temporal white matter. (C) Brain MRI three months after treatment onset. Flair weighted axial images (TR 10 002 ms; TE 160 ms) showing dramatic improvement under treatment. Hyperintense signal has diminished in both occipital lobes, but cerebral atrophy had appeared in the left occipital and temporal cortex. TE, time of echo; TR, time of repetition.

major role in the control of PML development; for example, in one study[5] no specific T cell response was demonstrated in a series of 14 patients before treatment, whereas nine of 10 survivors recovered specific immunity. Our patient thus probably had no specific T cell response against JCV before treatment, but recovered a moderate but significant response while on treatment, possibly explaining PML regression. Such a restoration of T cell response can be achieved by HAART in AIDS. This observation suggests that the combination of aracytine and cidofovir could have had a similar action in restoring a specific T cell response against JCV in our patient. A direct effect of these nucleosidic analogues against JC virus DNA may also explain the rapid clinical and radiological improvement in our patient.

Despite its substantial bone marrow toxicity, this observation suggests that the new association of intrathecal and intravenous aracytine with intravenous cidofovir could be useful in patients with PML, particularly those with an underlying haematological disease. It is worth noting that bone marrow toxicity did not lead to deterioration of the neurological status of the patient, supporting the view that a specific defect in CD4 function is more important than the absolute CD4 count. The dramatic improvement observed in our patient warrants further prospective studies testing this drug combination.

## Acknowledgements

We thank Dr Yacine Taoufik, Department of Immunology, Kremlin-Bicêtre Hospital, for his help with the immunological data and critical discussion.

**K Auré, A Béhin, F Louillet, C Lafitte, M Sanson**
Service de Neurologie Mazarin, Groupe Hospitalier Pitié-Salpêtrière, Assistance Publique-Hôpitaux de Paris, Paris, France

**J P Vernant**
Service d'Hématologie, Groupe Hospitalier Pitié-Salpêtrière

Correspondence to: Dr Anthony Béhin, Federation de Neurologie-Mazarin, Groupe hospitalier Pitié-Salpêtrière, 47-83 Boulevard de l'hôpital, 75013 Paris Cedex, France; anthony.behin@psl.ap-hop-paris.fr

doi: 10.1136/jnnp.2004.061408

Competing interests: none declared

## References

1 **Gasnault J**, Taoufik Y, Goujard C, *et al*. Prolonged survival without neurological improvement in patients with AIDS-related progressive multifocal leukoencephalopathy on potent combined antiretroviral therapy. *J Neurovirol* 1999;**5**:421–9.
2 **Aksamit AJ**. Treatment of non-AIDS progressive multifocal leukoencephalopathy with cytosine arabinoside. *J Neurovirol* 2001;**7**:386–90.
3 **Herrlinger U**, Schwarzler F, Beck R, *et al*. Progressive multifocal leukoencephalopathy: cidofovir therapy in three patients with underlying hematological disease [letter]. *J Neurol* 2003;**250**:612–14.
4 **Hall CD**, Dafni U, Simpson D, *et al*. Failure of cytarabine in progressive multifocal leukoencephalopathy associated with human immunodeficiency virus infection. *N Engl J Med* 1998;**338**:1345–51.
5 **Gasnault J**, Kahraman M, de Goer de Herve MG, *et al*. Critical role of JC virus-specific CD4 T-cell responses in preventing progressive multifocal leukoencephalopathy. *AIDS* 2003;**17**:1443–9.

# Diaphragmatic paralysis and respiratory failure as a complication of Lyme disease

There have been five recorded cases of diaphragmatic paralysis as a complication of neuroborreliosis.[1–5] Here we report another case of Lyme meningoradiculitis, caused by an identified tick, leading to bilateral diaphragmatic paralysis with an abbreviated course on treatment.

## Case report

A 59 year old female presented with a recent history of abdominal pain and falls because of a weakness in her right leg. She had been complaining of flu-like symptoms with twitches in her back and pain in her right side for a month. She gave a history of recently having been bitten by ticks whilst gardening. There was no history of any recent rash. On the day of presentation, she complained of a mild cough, reduced appetite, abdominal distension, constipation, and dysuria. She was a lifelong smoker but was generally healthy.

At presentation her blood pressure was 206/107 mm Hg. There was some epigastric tenderness. She had bruising on her right leg that she associated with the falls.

The chest radiograph on admission was unremarkable. Abdominal *x* ray showed dilated loops of small bowel and a loaded colon. Her only blood abnormality was hyponatraemia at 121 mmol/l. She was admitted for further investigations.

On day 3 of admission she became increasingly short of breath and on examination had decreased bibasal air entry. On day 4 her respiratory rate was 25/min and arterial blood gases (ABG) demonstrated hypoxaemia but adequate ventilation with pH 7.51, $Po_2$ 6.7 kPa, and $Pco_2$ 4.7 kPa. Her chest radiograph showed left basal changes. On day 5 her $Pco_2$ had risen to 6.8; she was admitted to the intensive care unit and non-invasive ventilatory support was commenced. She had a decreased inspiratory pressure and a decreased vital capacity. She was noted to have absent gag reflex and poor swallow and on day 6 was intubated to protect against aspiration pneumonia. The patient remained fully conscious and co-operative, easily triggering the ventilator but requiring significant inspiratory pressure support of 20 cm $H_2O$.

Neurological examination demonstrated right hip and knee extensor weakness (2/5), absent right knee jerk, and a loss of sensation on her left lateral thigh. Because she lived in a known endemic area we thought about Lyme disease, but we also considered differential diagnoses such as Guillain-Barre syndrome, listeriosis, and acute poliomyelitis. We commenced treatment with doxycycline whilst awaiting the results of further investigations. Around this time the patient indicated a small black lesion on her upper abdomen that was removed and on closer examination was identified as tick mouthparts (fig 1).

**Figure 1** Photomicrograph of tick recovered from patient, showing tick mouthparts.

Given her smoking history and persistent hyponatraemia, a chest computerised tomography was performed which showed only left lower lobe collapse and a small left pleural effusion. Bronchoscopy was unremarkable. Chest ultrasound screening showed only minimal movement of both diaphragms.

Cerebrospinal fluid analysis (CSF) demonstrated a white cell count 181 cells/mm$^3$ (100% mononuclear), red cell count 22 cells/mm$^3$, glucose 2.3 mmol/l (serum glucose 6.8 mmol/l), and protein 0.96 g/l. Immunological analysis of the CSF was not done because an insufficient sample was obtained. Brain magnetic resonance imaging (MRI) was normal. Spinal MRI and electromyography were not carried out. Immunoglobulin M and immunoglobulin G antibodies to *Borrelia burgdorferi* were detected in serum, and at this point intravenous ceftriaxone (2 g for 30 days) was commenced.

Over the next 7 days her strength increased and a repeat ultrasound of the diaphragm on day 16 of admission showed marked improvement with the right dome moving normally and some residual left sided weakness associated with overlying lung consolidation.

The patient was weaned from the ventilator and extubated following a total of 22 days of respiratory support. She underwent intensive physiotherapy and has made an uneventful recovery. One year later she complained of mild shortness of breath on lying flat with an exercise tolerance of 1 mile on the flat. On examination she had hyperaesthesia in her right leg with slightly brisk knee reflexes (previously absent right knee jerk). She had persistent bilateral diaphragmatic paralysis demonstrated on chest ultrasound screening. Pulmonary function tests showed a 40% reduction between erect and supine vital capacity measurements.

## Discussion

The first case of diaphragmatic paralysis as a complication of Lyme disease was reported in 1986: a 73 year old male, treated with ampicillin and netilmicin, who required ventilation for 3 months and then died after receiving treatment for a pulmonary embolism.[1] Another four cases have been reported in patients between the ages of 39 and 68, all of whom were treated with either doxycycline or ceftriaxone and two of whom required ventilation due to respiratory failure.[1–5] All patients were well at follow up, although one patient had persistent phrenic paralysis 6 months after treatment.[5]

In all previous cases of diaphragmatic palsy as a complication of Lyme disease, either the patient reported dyspnoea or hypoxia was noted on ABG. The diagnosis of phrenic nerve palsy was made by the following methods: hemidiaphragm elevation, fluoroscopic screening of diaphragmatic movements, or electrical stimulation of phrenic nerves.[1–5] Our patient had a lymphocytic meningitis with sensory and motor neuropathies including bilateral phrenic nerve palsies. Diaphragmatic paralysis due to Lyme disease was diagnosed on the basis of clinical features, chest ultrasonography, the presence of the tick head, and serology indicating a recent infection with *B burgdorferi* as well as a rapid response to antibiotic therapy.

The clinical diagnosis of Lyme disease may be supported by serologic testing. *B burgdorferi* antibody tests may be negative in early infection, but patients are usually seropositive at or shortly after presenting with neurological symptoms. In some patients, antibodies against *B burgdorferi* may be detectable in CSF slightly earlier than serum. Culture and *B burgdorferi* deoxyribonucleic acid detection using polymerase chain reaction may also be used but were not in our case.

The three patients reported in the literature with respiratory failure caused by neuroborreliosis were ventilated for 3 months, 1 month, and 13 months, respectively, whilst our patient required ventilation for only 22 days.[1 2 4] We speculate that early recognition of the possibility of Lyme disease and appropriate treatment shortened our patient's acute illness.

In conclusion, it is important to consider Lyme disease in the differential diagnosis of acute respiratory failure – with or without erythema migrans.

## Acknowledgements

We would like to thank Mr P R Randell of the Microbiology Department, St Richard's Hospital, Chichester, UK and Dr Susan O'Connell of the Department of Microbiology and Public Health Laboratory, Southampton General Hospital, Southampton, UK.

**R A Abbott, S Hammans, M Margarson, B M Aji**
St Richard's Hospital, Chichester, UK

Correspondence to: Rachel A Abbott, St Richard's Hospital, Spitalfield Lane, Chichester, West Sussex PO19 6SE, UK; rachel.abbott@nhs.net

doi: 10.1136/jnnp.2004.046284

Competing interests: none declared

## References

1 **Melet M**, Gerard A, Voiriot P, *et al*. Méningoradiculonévrite mortelle au cours d'une maladie de Lyme. *Presse Med* 1986;**5**:2075.
2 **Sigler S**, Kershaw P, Scheuch R, *et al*. Rspiratory failure due to Lyme meningoradiculitis. *Am J Med* 1997;**103**:544–547.
3 **Faul JL**, Ruoss S, Doyle RL, *et al*. Diaphragmatic paralysis due to Lyme disease. *Eur Respir J* 1999;**13**:700–702.
4 **Winterholler M**, Erbguth FJ. Tick bite induced respiratory failure. *Intensive Care Med* 2001;**27**:1095.
5 **Gomez de la Torre R**, Suarez del Villar R, Alvarez Carreno F, *et al*. Parálisis diafragmática y artromialgia por enfermedad de Lyme. *An Med Interna (Madrid)* 2003;**20**:47–9.

# Benign paroxysmal positional vertigo (BPPV) predominantly affects the right labyrinth

We read with great interest the article ''Benign paroxysmal positional vertigo predominantly affects the right labyrinth'', by M von Brevern *et al*,[1] which prompted us to review our data of the last 10 years (1995–2004).

A total of 661 patients, referred to the ear, nose, and throat department or to the neurology department, were diagnosed as having benign paroxysmal positional vertigo (BPPV) in its various forms. The pathology was located in the posterior canal in 477 patients, in the horizontal canal in 142, and in the anterior canal in 22. Multiple canals were affected in 20 patients (table 1).

The right ear was 1.50 times more frequently involved than the left. The predominance of the right ear was seen in all types of BPPV (table 1).

Hence, our data confirm the preponderance of right sided BPPV. The predilection of right sided BPPV was seen in all variants. Horizontal canal BPPV was observed in 22%, confirming our previous data.[2] This number is higher than in other series.[3 4] The apogeotropic form should not be considered as a rarity as it contributes to 23% of horizontal canal BPPV in our series and 38% in the series of Casani *et al*.[3]

**W Damman, R Kuhweide**
Ear, Nose and Throat Department, AZ St Jan, B-8000 Brugge, Belgium

**I Dehaene**
Department of Neurology, AZ St Jan, B-8000 Brugge, Belgium

**Table 1** Laterality of the affected ear in different forms of BPPV

| | | Horizontal canal | | | | |
|---|---|---|---|---|---|---|
| No. of patients | Posterior canal | Geotropic | Apogeotropic | Anterior canal | Multiple canals | Totals |
| Right n = 379 | 266 | 61 | 23 | 15 | 14 | 57% |
| Left n = 253 | 185 | 48 | 10 | 7 | 3 | 38% |
| Bilateral n = 29 | 26 | 0 | 0 | 0 | 3 | 5% |
| | 72% | 22% | | 3% | 3% | 100% |
| | | 77% geotropic | 23% apogeotropic | | | |

Total n = 661.